Extrafollicular Cystic Adenomatoid Odontogenic Tumor of the Maxilla: a Rare Challenging Case Report with Review of the Literature

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Abstract

Adenomatoid odontogenic tumor is a non-aggressive slow growing epithelial odontogenic tumor with varied clinical and microscopic features. This neoplasm rarely appears as a completely cystic lesion histopathologically; therefore, oral pathologists should be aware of the various histopathologic types of this lesion. The main objective of this case report is to present a rare challenging case of maxillary extrafollicular cystic adenomatoid odontogenic tumor affecting a 28-year-old male.

Keywords: odontogenic tumor, maxilla, benign, cyst, adenomatoid odontogenic tumor.

Introduction

Adenomatoid odontogenic tumor (AOT) is a benign slow growing epithelial odontogenic neoplasm representing 2% to 7% of all odontogenic tumors (1, 2). Ethnic differences have been reported on the prevalence of AOT, which show the lowest percentage in Europe and the highest in Africa (1, 3). It appears that dental lamina in the gubernacular cord of the developing permanent incisor, canine or premolar is the embryonic source of the majority of AOTs (4). It shows a predilection in the anterior portions of the maxilla and patients under 30 years (1, 3). The majority of cases demonstrate a well-circumscribed, unilocular radiolucency containing the crown of an unerupted tooth (follicular type) which mimic dentigerous cysts. However, an AOT encircles the crown as well as the root while dentigerous cysts envelop only the crown and is attached to the cemento-enamel junction (3). The extrafollicular variant is a well-delineated, unilocular radiolucency that is located between the roots of erupted teeth. The rare extraosseous type is also reported. Albeit, AOT may be entirely radiolucent or it could contain fine (snowflake) calcifications (1, 2, 4). It appears that the cut surface of the lesion is predominantly solid or may reveal some degrees of cystic change (2). In some cases, the solid portion may be present only as masses in the wall of a cyst (5, 6). Some studies have reported cases of purely cystic AOT (3, 5, 6). The aim of this case report is to present a rare case of extrafollicular cystic AOT affecting a 28 -year-old male.

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Case report

A 28-year-old man was referred to the Department of Oral and Maxillofacial Pathology, Shahid Beheshti University of Medical Sciences, Tehran, Iran, for evaluation of the painless swelling of the right maxillary vestibule in the area of 13, 14 for eight months. The patient had no history of previous trauma or medical problems. Intraoral examination revealed buccal cortical expansion in the area of right maxillary canine and the first premolar. Additionally, the overlying mucosa was intact and pink in color and the panoramic radiograph indicated a well-defined corticated radiolucent lesion between roots of 13 & 14, measuring 2×2 cm. Displacement of the adjacent vital teeth was also evident (Fig. 1). There was no cervical lymphadenopathy and blood investigation was within normal limits. Aspiration of the lesion showed a serous-like fluid. Due to radiographic features, fluid aspiration and vitality tests, a developmental cystic lesion such as odontogenic keratocyst (OKC) or lateral periodontal cyst (LPC) were considered in the differential diagnosis. The lesion was completely excised under local anesthesia. The gross was a cystic creamy elastic tissue and the maximum wall thickness was 2mm. The histopathologic sections demonstrated a cystic lesion lined with thin, non-keratinized flattened epithelium with clear cytoplasm (Fig. 2). Along with some area of the cystic wall, there was a proliferation of clear and spindle cells admixed with dentinoid material, cementum-like and dystrophic calcification (Fig. 3). A distinctive diagnosis based on these microscopic features and the radiographic image was impossible; thereby, the serial sections of the paraffin block were carried out. Interestingly, some epithelial islands composed of spindle cells were found that formed whorled masses of cells and duct-like structures in the fibrous wall of the cyst within deep sections. Inductive change of the connective tissue around the islands was evident (Fig. 4). According to these findings, the diagnosis of an extrafollicular cystic AOT was performed. Consequently, clinical follow-up was done after conservative surgery and the patient was free of tumor for 1 year postoperatively.

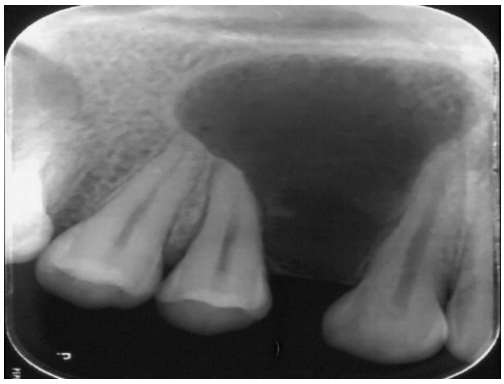

Figure1. The periapical radiograph revealed a well-defined corticated unilocular radiolucent lesion with root divergence of 13, 14

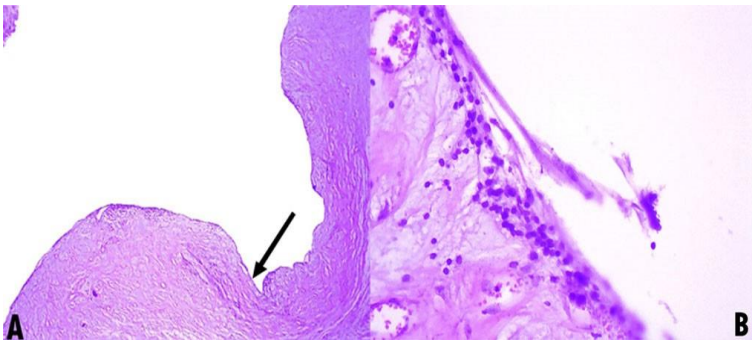

Figure2. A) Microscopic sections show a cystic lesion lined by thin non-keratinized flattened epithelium (black arrow) (hematoxylin-eosin stain, original magnification×40). **B)** Thin cystic lining with clear cytoplasm (×400)

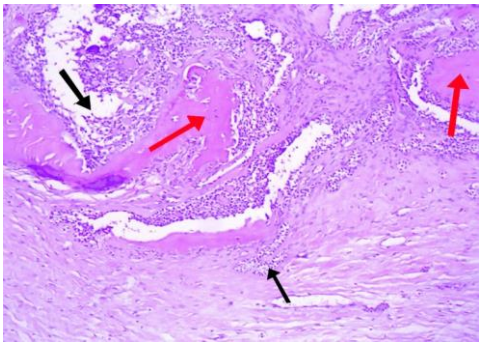

Figure3. Nests and sheets of clear cells (black arrow) admixed with large masses of dentinoid material (red arrow) (×100).

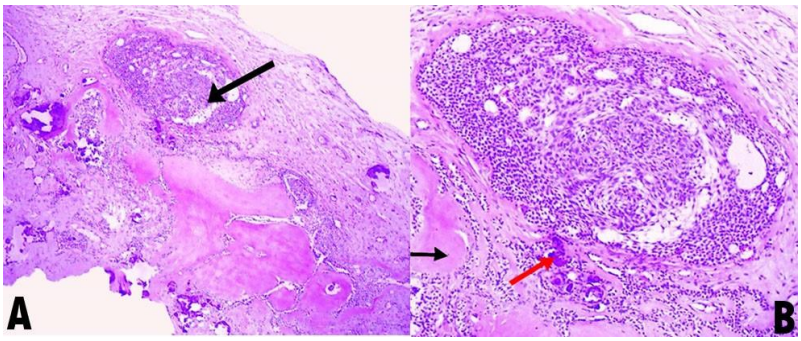

Figure4. A) Fibrous wall of the cyst shows spindle cells that formed whorled masses of cells and duct-like structures (black arrow). Dentinoid and cementum-like materials are also seen (×100).
B) island of AOT, dentinoid (black arrow) and cementum-like material (red arrow) (×400).

Discussion

AOT has a high tendency toward women in the second decade of life (6). Our case was male in his 3th decade of life. A number of these lesions show remarkable growths that support the classification of AOT as a benign tumor and not a hamartoma (1). AOT is commonly asymptomatic and is discovered during routine radiographic examinations. Larger lesions show painless expansion of the jaw (1, 2). The present case also had expansion. AOT is usually surrounded by a thick, fibrous capsule. It is composed of spindle shaped epithelial cells that form sheets, strands, whorled masses, rosettelike or duct like structures in a fibrous connective tissue. Abortive enamel formation, dentinoid material and cementum may also be scattered throughout the neoplasm (2). It shows a lot of diverse microscopic features such as clear cell changes, calcifying epithelial odontogenic tumor (CEOT) like areas or pigmentation (7). AOT is rarely seen as completely cystic in the microscopic feature. Some authors have suggested ‘‘Adenomatoid Odontogenic Cyst’’ (AOC) to be a more appropriate term and discuss that the lesion is a cyst with intraluminal proliferation which fills the cystic space giving a solid appearance (5). In addition, Kumar et al (3) claimed that AOT is not a tumor, but a cyst that exhibits a hamartomatous intraluminal proliferation of epithelial cells derived from the Hertwig epithelial root sheath. In contrast, Thakur et al (8) support the tumoral nature of this lesion. In the end, Philipsen, who has carried out copious numbers of empirical studies on this matter, prefers the name “AOT” rather than “AOC” (4). AOT is rarely seen in association with other benign odontogenic neoplasms or cysts (6). Dentigerous cyst, calcifying odontogenic cyst, unicystic ameloblastoma and odontoma have all been reported as components of AOT (9-14); several cases of de novo cystic AOT have also been reported (3, 5, 6, 15). In all of these cases, the lesion was associated with an impacted tooth radiographically, while our patient indicated an extrafollicular type. In addition, in the reported cases, there had been a male predilection and their age ranged from 12 to 19 years. Gadewar et al (5) found that very few case reports of cystic AOT have described a cystic lining. This lining is defined as thin, non-keratinized stratified squamous epithelium, with or without nodule formation, evidence of calcification in cyst stroma and subepithelial hyalinization, resembling dentinoid material. In our case, the cyst was lined with thin nonkeratinized epithelial cells with clear cytoplasm and no evidence of nodular thickening or subepithelial hyalinization. Conversely,

large amounts of dentinoid and cementum-like materials was evident in fibrous stroma. The treatment of choice in all variants (AOT, AOC and extraosseous) is conservative surgery with no evidence of recurrence (2, 5). Reported cases of AOT with recurrence and aggressive behavior appear to be related to another destructive tumor called adenoid ameloblastoma (16). This neoplasm reveals distinct microscopic structures reminiscent of AOT and ameloblastoma, along with hard tissue (dentinoid) formation and an aggressive behavior (17).

In conclusion, AOT may resemble an odontogenic cyst; therefore, oral pathologists should be aware of the various microscopic features of this lesion. Moreover, when the microscopic examination of the odontogenic lesion is not diagnostic, obtaining serial sections of the lesion is highly recommended.

Conflict of interests

The authors declare that there is no conflict of interests regarding the publication of this paper.

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